Results of Surgical Correction of Anorectal Malformations

A 10–30 Year Follow-up

NAOMI IWAI, M.D., JUN YANAGIHARA, M.D., KAZUAKI TOKIWA, M.D., EIICHI DEGUCHI, M.D., and TOSHIO TAKAHASHI, M.D.

From the Division of Surgery, Children's Research Hospital, and the First Department of Surgery, Kyoto Prefectural University of Medicine, Kyoto, Japan

Of 119 patients with surgical correction of anorectal malformations, 47 who were 10–30 years of age were interviewed personally and had manometric studies to evaluate postoperative continence. This clinical study included not only long-term anorectal function but also sexual function. Patients with low type anomalies or with intermediate type anomalies were more likely to be continent, whereas patients with high type lesions had some problems with continence. However, only two of the 16 patients (12%) with high type anomalies were classified as having poor results. This rate is perhaps lower than might be expected. Thus, incontinent patients may become continent even if they were classified as having fecal incontinence before 6 years of age. Most patients who were 15–30 years of age had normal sexual function except for two females with irregular menstruation. These results indicate that achievement of fecal continence and sexual function in patients with high type anomalies treated by abdominoperineal rectoplasty depends on careful dissection as close as possible to the rectal wall and bringing the terminal bowel down exactly within the sling of the puborectal muscle.

THE MAIN OBJECT OF any surgery for anorectal malformations is to achieve anal continence. In addition to bowel function, sexual function is also important. However, very few reports[1,2] have mentioned sexual function after surgery for anorectal malformations.

This paper reports the long-term results with regard to bowel and sexual functions in patients treated surgically for anorectal malformations who are now 10–30 years of age.

Clinical Data

One hundred forty-one patients with anorectal malformations were treated at the Children's Research Hospital of Kyoto Prefectural University of Medicine from 1960 to 1986 (Table 1). Of these patients, 66 (51 males and 15 females) had high type anomalies, 24 (16 males and 8 females) had intermediate type anomalies, and 51 (34 males and 17 females) had low type anomalies.

A total of 121 surgical corrections were performed on 119 patients (Table 2). Two of the 121 patients had undergone cut-back anoplasty in the neonatal period and anal transplant at 2 or 3 years of age.

The usual operative procedure in this division has been colostomy for the high and intermediate types in the neonatal period, followed by abdominoperineal rectoplasty, in which the rectum was dissected carefully along the rectal wall to preserve the pelvic nerves. The sling of the puborectal muscle was well identified by causing contraction of the puborectal muscle with an electric stimulator, and the pelvic floor was pushed upward from the perineal side by the surgeon's left hand so that the sling of the puborectal muscle could be well visualized. The center of the external sphincter muscle was also identified by causing contraction of the muscle with the electric stimulator. Low type anomalies were treated by neonatal perineoplasty. Anal transplants were performed at 3 or 4 months of age after anal dilatation, or cut-back anoplasty was performed soon after birth, followed by an anal transplant at 2–3 years of age.

Postoperative complications (Table 3) were observed in 31 patients (26%). Prolapse of the rectal mucosa occurred in 14 patients, and the excess rectal mucosa required trimming in all 14 patients. Anal stricture was present in eight patients, leading to secondary megacolon in two patients. Three of the eight patients required

Reprint requests: Naomi Iwai, M.D., Division of Surgery, Children's Research Hospital, Kyoto Prefectural University of Medicine, Kamigyo-ku, Kyoto, 602, Japan.

Submitted for publication: August 14, 1987.

TABLE 1. *Types of Anorectal Malformations (1960–1986)*

Malformations in Male Patients	No. of Cases	Malformations in Female Patients	No. of Cases
High			
Rectourethral fistula	36	Rectovaginal fistula	7
Anorectal agenesis without fistula	7	Rectovesical fistula	1
Rectovescial fistula	4	Rectocloacal fistula	3
Cloacal extrophy	3	Cloacal extrophy	2
Unknown	1	Unknown	2
Total	51 (13 deaths)		15 (5 deaths)
Intermediate			
Anal agenesis without fistula	9	Anal agenesis without fistula	1
Rectobulbar fistula	6	Rectovestibular fistula	3
Anorectal stenosis	1	Rectovaginal fistula (low)	3
		Anorectal stenosis	1
Total	16		8 (1 death)
Low			
Anocutaneous fistula	26	Anovestibular fistula	10
Covered anus–complete	8	Anovulvar fistula	5
		Covered anus–complete	1
		Perineal canal	1
Total	34		17

secondary anoplasty since anal dilatation was not effective. Intestinal obstruction due to adhesions was present in four patients after abdominoperineal rectoplasty, and all four patients recovered after surgical division of the adhesions. Bowel retraction after pull-through occurred in two patients and recurrent rectovaginal fistula occurred in one patient.

TABLE 2. *Surgical Corrections in 119 Patients with Anorectal Malformations*

Surgical Correction	No. of Patients		
	Male	Female	Total
Staged abdominoperineal rectoplasty	49	11	60 (50%)
Anal transplant	—	10 (2)	10 (8%)
Perineoplasty: perineal anoplasty	6	2	8 (6%)
Cut-back anoplasty	31	12	43 (36%)

TABLE 3. *Complications of Treatment in 119 Patients with Anorectal Malformations*

Complications	No. of Cases
Prolapse of rectal mucosa	14 (12%)
Anal stricture	8 (6%)
Intestinal obstruction	4 (3%)
Secondary megacolon	2 (2%)
Bowel retraction after pull-through	2 (2%)
Recurrent rectovaginal fistula	1 (1%)
Total	31 (26%)

TABLE 4. *Types of Anomaly and Age at Time of Follow-up Assessment*

Type of Anomaly	No. of Cases	Age (Years)	(Mean ages)
High type	16	10–20	(12)
Intermediate type	13	10–24	(13)
Low type	18	10–30	(13)

Current Bowel Control

Forty-seven patients, 10–30 years of age, were interviewed personally and assessed clinically: 16 patients, 10–20 years of age, with high type anomalies, 13 patients, 10–24 years of age, with intermediate type anomalies, and 18 patients, 10–30 years of age, with low type anomalies (Table 4).

Function was evaluated by the Kelly score system[3] on the basis of three criteria: (1) control of feces and bowel habits, (2) fecal staining, and (3) sling action of the puborectal muscle. The results were classified as good (Kelly 5–6), fair (Kelly 3–4), and poor (Kelly 0–2).

The current bowel control of these patients is shown in Table 5. All 18 patients with low type anomalies had good control, and of the 13 patients with intermediate type anomalies, 10 (77%) achieved good control. On the other hand, only seven of the 16 patients with high type anomalies had good control, and two of them (12%) had poor control.

Manometric assessment was usually performed without anesthesia, except in restless children who required mild sedation. The probe was perfused with a constant infusion of 10 ml/h. Of the 47 patients assessed clinically, 41 had manometric studies to evaluate postoperative continence. The results are summarized in Table 6.

All of the 14 patients with low type anomalies had both a high pressure zone in the anal canal and an anorectal reflex. Of the 12 patients with intermediate type

anomalies, 11 (92%) had a high pressure zone and six (50%) had an anorectal reflex. Of the 15 patients with high type anomalies, however, 11 (73%) had a high pressure zone and only three (20%) had an anorectal reflex.

Sexual Function (Tables 7 and 8)

Five male patients, 15–20 years of age, were interviewed regarding their sexual function: erection, ejaculation, and history of marriage. All five patients had normal erections and ejaculations. One patient was married and enjoyed normal sexual activities.

Seven female patients, 15–30 years of age, were interviewed regarding menstruation and history of marriage. Five of the seven had regular menstruation, and two had irregular menstrual periods; one had had a rectocloacal fistula and the other had a rectovaginal fistula. Three patients were married, and one had had a normal pregnancy and delivery.

TABLE 5. *Clinical Assessment and Type of Anorectal Malformation*

Type of Anomaly	No. of Cases	Clinical Assessment		
		Good	Fair	Poor
High	16	7 (44%)	7 (44%)	2 (12%)
Intermediate	13	10 (77%)	3 (23%)	0
Low	18	18 (100%)	0	0

TABLE 6. *Results of Anorectal Manometry*

Type of Anomaly	No. of Cases	Anorectal Manometry	
		Presence of HPZ	Presence of Anorectal Reflex
High	15	11 (73%)	3 (20%)
Intermediate	12	11 (92%)	6 (50%)
Low	14	14 (100%)	14 (100%)

HPZ = high pressure zone.

Discussion

Specific complications of surgery were observed in 31 of 119 (26%) patients who had had surgical treatment for anorectal malformations. This rate is somewhat lower than that in series of Hecker et al.[4] Anal strictures were especially less frequent, presumably because of careful anal dilatation begun 2 weeks after operation. On the contrary, the rate of mucosal prolapse in our series was 12%, which is almost three times that in the series of Hecker et al. The goal of a cosmetically satisfactory anal opening as well as one that functions normally depends primarily on the operative technique. Therefore, care should be taken to slide the perianal skin flap into the new anal canal so that mucosal prolapse is avoided.

In the current series poor results were not observed either in patients with intermediate type anomalies or in those with low type anomalies. However, two of the 16 patients (12%) with high type anomalies had poor results, and the remaining 88% had good or fair results. In our previous report,[5] 23% of those with high type anomalies had poor results, but the mean follow-up period was 5.75 years. Therefore, this repeat clinical assessment demonstrates that incontinent patients may become continent even if they had fecal incontinence before 6 years of age. Smith et al.[6] stressed the importance of parental assistance in training and of the patient's desire, cooperation, and mental ability. Intellectual development after six years of age through the experiences of school life is one of the steps towards the achievement of continence, in addition to physiotherapy.

We previously reported that continent patients characteristically had a very high pressure zone in the anal canal. Therefore, in view of the current manometric evidence, a high pressure zone and an anorectal reflex were considered to be chief parameters for objective assessment. A high pressure zone was present in 73% of the high type anomalies. This finding correlates well with the clinical evaluation, in which 88% of the patients with high type anomalies had good or fair results. On the other hand, only 20% of the patients with high type anomalies had an anorectal reflex. In our previous study, 20% of the patients with high type anomalies had an anorectal reflex. These results suggest that high pressure zones might be achieved in patients with good or fair results by contractions of the voluntary muscles as intellectual development progresses or as a result of physiotherapeutic training.

TABLE 7. *Sexual Function in Male Patients*

Case	Age (Years)	Type of Anomaly	Erection	Ejaculation	Marital Status
1	18	High	Normal potency	Normal	Single
2	15	High	Normal potency	Normal	Single
3	20	High	Normal potency	Normal	Married
4	17	Intermediate	Normal potency	Normal	Single
5	20	Low	Normal potency	Normal	Single

TABLE 8. *Sexual Function in Female Patients*

Case	Age (Years)	Type of Anomaly	Menstruation	Marital Status
1	17	High	Irregular	Single
2	15	High	Regular	Single
3	17	Intermediate	Irregular	Single
4	24	Intermediate	Regular	Married
5	16	Low	Regular	Single
6	25	Low	Regular	Married (1 child)
7	30	Low	Regular	Married

Sexual dysfunction after major rectal surgery in adults is extremely common.[7,8] In the current study, however, most of the patients with anorectal malformations had normal sexual activity except for the two patients with irregular menstruation. Nixon and Puri also reported that all patients examined had normal sexual activity except for one patient with a permanent colostomy.[1] These results indicate that in the surgery of anorectal malformations, refinements of surgical technique, such as dissection as close as possible to the rectal wall, can prevent postoperative sexual disturbances. Therefore, the majority of patients with anorectal malformations can expect a good quality of sexual activity.

References

1. Nixon HH, Puri P. The results of treatment of anorectal anomalies: a thirteen to twenty year follow-up. J Pediatr Surg 1977; 12:27–37.
2. Ishihara M, Morita K. Long term results of surgical treatment of imperforate anus (in Japanese). Jpn J Pediatr Surg 1979; 11:661–667.
3. Kelly JH. Cineradiography in anorectal malformations. J Pediatr Surg 1969; 4:538–546.
4. Hecker WC, Holschneider AM, Kraeft H, Neuman M. Complications, lethality and long-term results after surgery of anorectal atresia. Z Kinderchir 1980; 29:238–244.
5. Iwai N, Ogita S, Kida M, et al. A clinical and manometric correlation for assessment of postoperative continence in imperforate anus. J Pediatr Surg 1979; 14:538–543.
6. Smith EI, Tunel WP, Williams GR. A clinical evaluation of the surgical treatment of anorectal malformations (imperforate anus). Ann Surg 1978; 187:583–592.
7. Danzi M, Ferulano GP, Abate S, Galifano G. Male sexual function after abdominoperineal resection for rectal cancer. Dis Colon Rectum 1985; 26:665–668.
8. Bulslev I, Harling H. Sexual dysfunction following operation for carcinoma of the rectum. Dis Colon Rectum 1985; 26:785–788.